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On Cystic and Encysted Solid Tumours of the Jaws,

WITH OBSERVATIONS ON THE STRUCTURE OF
THE ENAMEL ORGAN.

BY

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MR. PRESIDENT AND GENTLEMEN,

It was with much pleasure that I acceded to the request of your Secretary that I should read a paper before this Society, knowing that I should have the honour of addressing gentlemen having an intimate knowledge of the subject I have chosen, although regarding it from a somewhat different standpoint to myself. Tumours, particularly cystic tumours, of the jaws have for some time attracted my attention, and they formed the subject of a lecture* which was published in the *British Medical Journal* in January, 1883. It is to some of the views therein brought forward that I desire, in part, to ask your indulgent attention, and I need hardly apologise, seeing the extent of contemporary medical literature, for passing to some extent over old ground, more

* An Erasmus Wilson lecture delivered at the Royal College of Surgeons, 1882.

especially as the conditions of the present meeting enable me to illustrate the subject by microscopic drawings and specimens.

It is my intention neither to speak of the single or simple cysts met with in the jaws—namely, the small inflammatory cysts attached to the roots of teeth, which Magitôt has designated as periosteal cysts,—nor of cysts directly connected with more or less perfectly developed tooth-follicles, which are included under the general names of dentigerous or follicular cysts. But, in the first place, I shall enter into the consideration of certain points in the pathology of the multilocular cystic epithelial tumour (formerly known as multilocular cystic disease, cystic or adeno-sarcoma) that appear to me to have a more immediate bearing on your speciality; it will, however, be necessary to prelude my observations by a brief description of the clinical character of these tumours.

This disease occurs at all periods of life from infancy to old age, but is commonest in early adult or middle age; of twenty-six cases which I collected, twenty patients had reached the age of twenty years when they came under observation. It more commonly affects the lower than the upper jaw, and the molar region, as in the odontomata, is usually, but not exclusively, involved. Its onset is insidious and its progress slow, extending in some cases over a period of twenty

years. The patient notices a swelling near the alveolus of a carious or inflamed tooth, or a blow has been received on the part, but in many cases no such exciting cause has existed. The swelling gradually increases, and the teeth overlying it become loosened and may fall out. Not infrequently a glairy fluid is discharged from a vacant alveolus, of which the orifice may become ulcerated.

In the lower jaw the morbid growth invades the cancellous tissue and expands the compact walls of the bone, chiefly the outer, giving rise to a prominence, which is at first rounded, but later, from the formation of numerous cysts, assumes a largely nodulated form. As its growth progresses the osseous walls become thinned, and in some parts yield to pressure with a parchment-like crackle, while in others they become absorbed and fluctuation may be obtained. In the upper jaw the growth projects into and distends the antrum, with at first little expansion of the outer surface; and the cyst formation is usually less marked.

A section of a typical specimen of the disease in the lower jaw displays an agglomeration of cysts, of sizes extending to an inch or more in diameter, and divided by thin septa of fibrous or, in some instances, of osseous tissue.

Most of the cysts are filled with a thick

glutinous fluid, and the larger often with a brown serous fluid. The advancing or younger portions of the tumour consist of a reddish-brown friable substance, not unlike, and which doubtless has often been mistaken for, a myeloid sarcoma. In some tumours the cysts are uniformly of small size.

I have shown that in minute structure these tumours are composed of branching and anastomosing rods or columns of epithelium, portions of which are cut off and form alveoli. The original germinal epithelium is modified in various degrees in different tumours, and in different parts of the same. The outer layer of cells, forming the columns and alveoli, become elongated or columnar; the central cells undergo colloid degeneration, and give rise more or less perfectly to the appearance of a reticulum of stellate cells, thus reproducing the structure of the rudimentary enamel organ.

The scanty stroma is composed of fibrous tissues, but when abundant consists of embryonic tissue in various stages of development towards the formation of fibrous tissue.

The colloid degeneration of the cells, and the accumulation of fluid in the alveoli, gives rise to the formation of cysts.

It is to the mode of degeneration of these cystic tumours that I wish to call your notice, not only on account of its pathological interest, but chiefly

because it appears to me materially to assist us in arriving at a conclusion in regard to a point in the histology of the development of the teeth which is not finally settled, namely, the mode of degeneration and removal of the central cells of the enamel organ.

Before discussing this question, let me in a few words recall to your minds the salient facts of the development of the teeth.

About the seventh week of embryonic life a downward projection or ingrowth of the epithelium lining the dental groove takes place along the whole length of the jaws. At certain points corresponding to the future teeth, papilla-like projections extend downwards from this ingrowth to form the rudimentary enamel organ. Coincident changes take place in the subjacent embryonic tissue resulting in the formation of a mass of gelatinous connective tissue, the tooth papilla, of which the round upper surface projects into and invaginates the rudimentary enamel organ, converting it into the enamel cap. The enamel organ is at first composed of round rudimentary epithelium, lined by a layer of elongated or sub-columnar cells, like to and continuous with those composing the deep layer of the epithelium of the mouth. In the progress of its development, the cells composing the enamel cap are differentiated. The inferior or inner layer—that nearest the tooth papilla—forms very

regular columnar cells, from which the enamel is subsequently developed; the outer layer remains for a time polyhedral; while the intermediate cells, or those composing the middle layer, undergo a peculiar transformation, resulting in the formation of a tissue composed, apparently, of stellate anastomosing cells, and formerly thought to be mucous connective tissue; this layer subsequently disappears. Two views may be entertained regarding the manner in which this peculiar change in the middle layer of cells is brought about: the one, which I must own is very generally accepted, and receives the support of the high authority of Dr. Klein,* is that the cells are compressed and flattened by an accumulation of fluid between them, that is, in the intercellular substance; the other, that the cell substance itself is degenerated, and this view is apparently held by Waldeyer, who says that he is unacquainted with a similar metamorphosis in any part of the body except in the epithelium of the Graafian follicles, "but never occurring [there] in as regular a manner."† He, however, does not enter into an explanation of the nature of the process.

Now I shall endeavour to show that the latter is the correct view, and that the metamorphosis in question is the result of a form of liquefaction

* Atlas of Histology.

† Sticker's "Handbook of Histology," vol. i. p. 280.

or colloid degeneration of the protoplasm. But, before arguing from pathological appearances in order to elucidate histological changes, it is necessary to substantiate more completely the fact that these tumour formations are structurally analogous to the rudimentary enamel organ. The resemblance was not only observed by myself in greater or less perfection in twelve specimens which I examined, but two cases have been also described in Germany by Falkson and Bryck respectively, and these so closely resembled the enamel organ, that, from this circumstance mainly, they argued that the growths proceeded from the rudiments of a tooth arrested in its development. Quite recently Bernays, an American surgeon, in describing a similar case, has adopted the same view.

It is true that the tumour formations present considerable differences among themselves, but the forms are all transitional, and grades from the most elemental to the most distinctly marked type of the rudimentary enamel organ, are even observable in the same tumour.

The mode of degeneration of the epithelium is more clearly evident in those tumours in which the cyst formation is well marked. The central cells of the columns and alveoli become much enlarged, finely granular, and the nucleus is pressed against the cell wall. Then the protoplasm of the

individual cells becomes merged into a general mass, while the nucleus and the portions of cell wall attached to it remain visible, and have somewhat the appearance of flattened cells with processes lying in the interstices of the degenerated cells. In other instances a drop of colloid substance forms within the cell, by which the protoplasm is expanded into a narrow band, and the nucleus being pressed to one side, the characteristic "signet-ring" form is produced. (See Fig. I.) The approximation of such cells with their nuclei at the points of contact will, as you may readily conceive, give rise to a wide mesh-work. In the layer intermediate between the central and peripheral portions of the columns, the degenerative change is less extensive and the resemblance to the central network of the enamel organ is exact (Figs. II, III, IV). Vacuoles form at one or more points in the slightly enlarged cells, sometimes separating the nucleus, which for the most part retains its rounded form, from the cell wall. A kind of solution of the protoplasm takes place, the vacuoles in adjoining cells run together, and finally the nuclei remain connected only by filaments of undissolved protoplasm and the vestiges of the cell walls. Further, in studying with high power immersion lenses some beautiful sections of the rudimentary enamel organ kindly lent me by Mr. Charles Stewart, I observed quite unmistakable "signet-



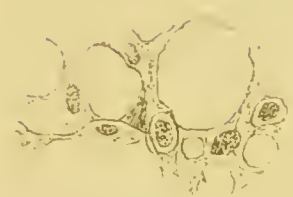
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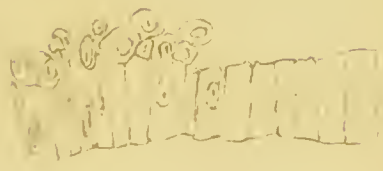
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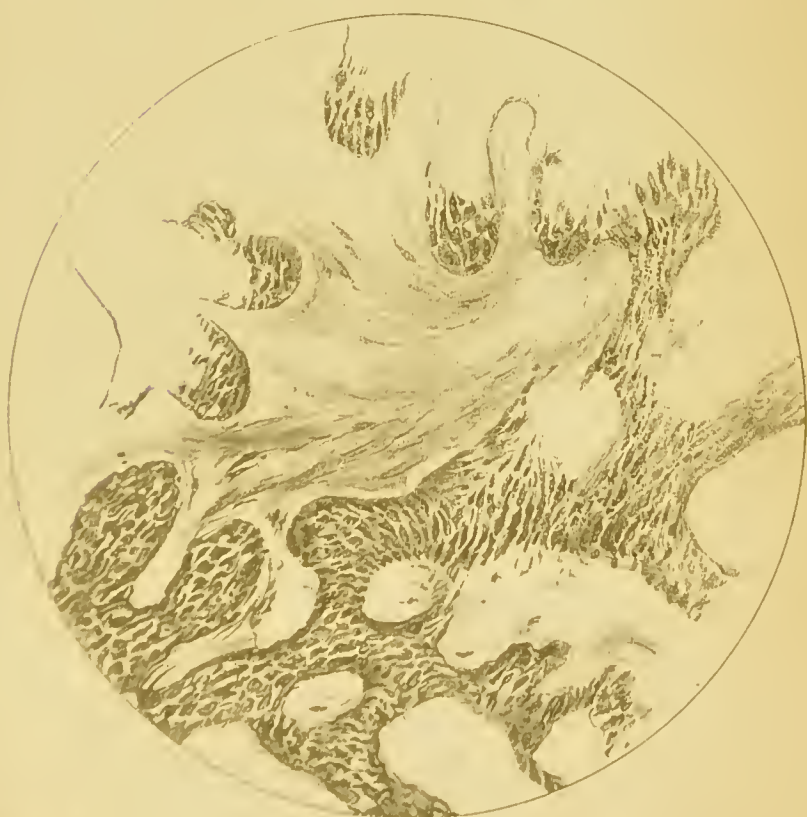
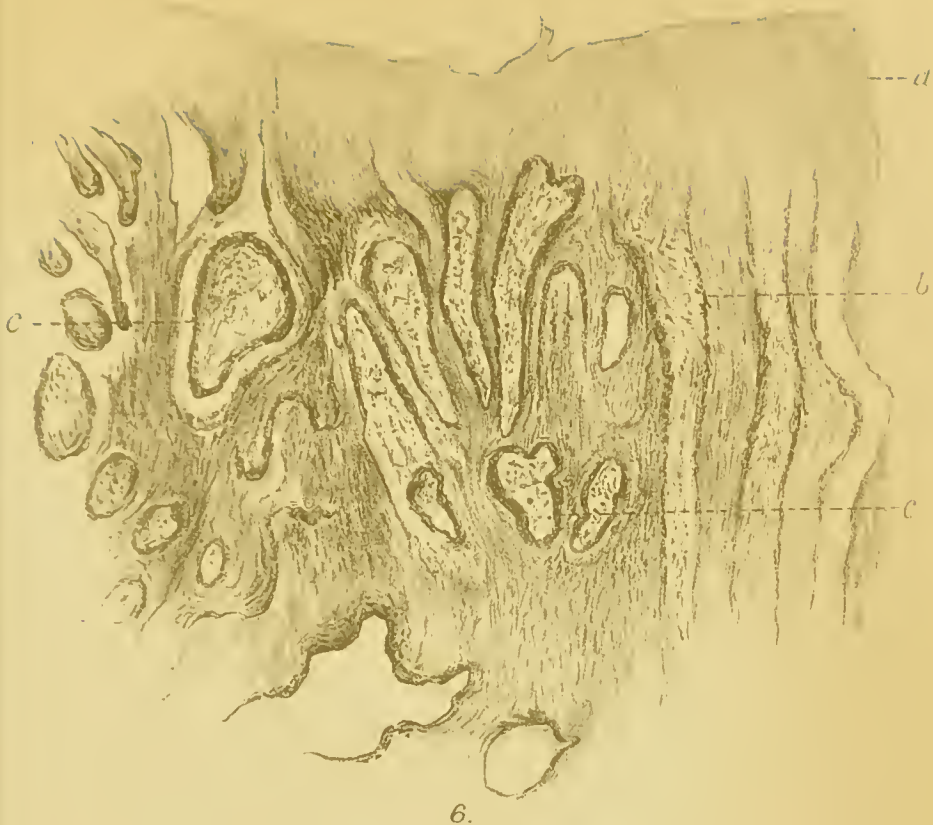
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b



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ring" cells (see Fig. V), and could in places make out a swelling with hyaline degeneration and vacuolation of the protoplasm prior to the formation of a network. If the epithelial cells were merely compressed by fluid accumulating between them, there would be no swelling of their substance, and in place of the indistinct fragment of the undissolved protoplasm around the nucleus, there would be a well-defined contour produced by the wall of the compressed cell.

Again, having already shown that the pathological products are structurally identical with the developing enamel organ, I contend that since the network in the former is clearly due to colloid degeneration of the cell substance, the same explanation also holds good for the latter; for unity of type is as clearly evident as regards mode of development and structure between pathological formations and normal tissues as between the lower and higher organisms of the animal kingdom, and unity of structure and form is the resultant of similar innate tendencies in the ultimate elements of the tissues, namely, in the cells themselves.

Before closing this portion of our subject, permit me to say a few words on the mode of origin of the multilocular cystic tumours. The inference naturally drawn from their structure would be, that they originated directly from the enamel

rudiment of an aborted ~~or supernumerary~~ tooth—in fact, that they are a form of embryo-plastic odontome. But of this there is no evidence, and the age at which they commonly occur is distinctly against such an assumption. Bryck is of opinion, but as I think on insufficient grounds, that the history in his case supported this conclusion.

In some two or three of the twelve specimens which I examined there was distinct evidence that the tumours proceeded from an ingrowth of epithelium continuous with that of the surface of the gum, and in two instances they occupied the margin of ulcerated alveoli from which teeth had fallen out. (See Fig. VI.) In another example there were flask-shaped masses of degenerating epithelium extending downwards from the gum between the teeth.

Since these observations were published M. Malassez communicated to the Société de Biologie of Paris* the result of the examination of a normal maxilla, carried out with the view of ascertaining if epithelial structures persisted in the neighbourhood of the alveoli, from which the cystic tumours of the jaws might originate. His search was rewarded by finding spherical, oval, and cylindrical masses of epithelium within the peri-odontal membrane (alveolo-dental ligament), and extending from the gum to the apex of the fang, but chiefly distributed

* "Journ. Soc. de la Biologie," 1884, p. 242.

below the neck and at the upper part of the root. These masses, he thinks, proceed from some of the epithelial formations accompanying dentition, probably from the external layer of the enamel organ. In this fact we have very strong confirmation of my observation above alluded to, and an explanation of the circumstances that these tumours usually begin near alveoli, soon lead to the extrusion of teeth, and that their interior often communicates with the mouth by an open alveolus.

In conclusion of this portion of our subject I beg to submit that the multilocular cystic tumours represent a distinct variety originating from epithelial ingrowths around the dental alveoli, possibly (if I may be allowed to indulge in a harmless speculation) in ingrowths the sole representatives of teeth long since suppressed in the process of evolution of our species, or from an overgrowth of the normal epithelium of the gum; a variety of tumour owing its peculiarities in structure and secondary metamorphosis to the inherent developmental peculiarities or tooth-forming properties of the epithelium from which it springs. It belongs to the group of epitheliomata, and possesses a decided, although not a high, degree of malignancy—facts which I endeavoured to prove in the lecture before alluded to. General dissemination of the tumour throughout the system may even take place, and the frequency of recurrence

is now placed beyond doubt by cases cited by Mr. Heath in the recent edition of his "Injuries and Diseases of the Jaws," a work which may almost be considered as the foundation of our knowledge of tumours of the jaws. In one of these cases the disease appeared on the cheek as "a typical epithelial ulcer."

Clinically resembling but differing pathogenetically from the multilocular cysts, is this, I believe, unique specimen of single cyst occupying the whole of the right side of a jaw.* It contains no trace of a tooth, and the structure of its wall shows that it is not dentigerous or follicular in the strict sense of the term, for I found that it was lined by a thick layer of small round epithelium. I have hazarded the conjecture that it may have originated from the expansion of a rudimentary enamel organ owing to the collection of fluid in its interior.

I will now with your permission pass on to the second part of my subject, and will bring to your notice one or two remarkable examples of encysted tumours of the jaw, that is, of solid tumours which, having expanded the walls of the jaw, are enclosed by a bony capsule. I need scarcely remind you that clinically there is at times considerable difficulty in distinguishing such tumours from cysts or cystic tumours. Their external

* Royal College of Surgeons' Museum, No. 2194.

appearances are often similar, in both the parchment-like crackling may be obtained, and, in many cases, a diagnosis is impossible without the aid of a puncture.

For these reasons, and from the fact that they may both be connected with anomalies in the development of the teeth, it may not be disadvantageous to place them in juxtaposition.

All the varieties of tumours of the jaws, with the exception of the osseous and cartilaginous, may be more or less definitely encapsuled or encysted. The fibrous and fibro-sarcomatous, sarcomatous, central myeloid tumours, odontomata, and lastly, solid epithelial tumours having no relation as regards their origin with the tubular carcinomata of the superior maxilla which spring from the epithelium and glands of the palate and antrum.

The specimens I am about to describe are at least closely related to, and should, I think, be classed among the odontomata. The odontomata, as you are well aware, were divided by Broca in his classical essay into odontomes embryoplastique, odonto-plastique, coronaire, and radicaire.

In the class of embryoplastic odontomata, Broca includes all encysted sarcomatous and fibrous tumours of the jaws, which he regards as the product of a tooth-rudiment in its earliest stage of development.

The odonto-plastic odontomata are tumours of which the tissues have reached a higher stage of evolution. They are either surrounded by a layer of odontoblasts, or have become more or less completely dentified. The distinguishing characters of the coronary and radicular odontomes do not concern our present subject.

CASE 1.—A very curious and, I believe, unique tumour came under my notice while I was Surgical Registrar in St. Bartholomew's Hospital. James B., aged twenty-four, was admitted to Henry Ward, under the care of Mr. Thos. Smith, with a swelling on the outer surface of the left side of the lower jaw. He first observed a prominence four years and four months previously, and for a year and a half before that had suffered pain in the part. For the first six months there was a discharge from the gum over the swelling. Eighteen months after the first appearance of the tumour two healthy teeth, situated in the affected part of the jaw, were extracted, and another at a later period.

The swelling had not increased during the last three years, and was not painful.

*Condition on Admission:—*A hard, rounded swelling, about three-quarters of an inch in diameter, projected from the outer surface of the lower jaw at the level of the molar teeth; the corresponding portion of the inner surface of the

jaw was slightly thickened. No crackling but slight and hardly perceptible yielding could be obtained by firm pressure over the tumour. A slit-like ulcerated aperture opened on the alveolar surface of the tumour, into which a probe could be passed for some distance. His dentition was very irregular, the full complement of teeth having only existed in the left superior and inferior maxillæ. In the right superior maxilla the canine and second molar, and in the right inferior maxilla the canine and first bicuspid, were absent.

The operation was delayed for a time on account of an attack of inflammation of the jaw, with much swelling of the cheek, which came on immediately after his admission.

Operation:—An incision having been made along the lower margin of the ramus, the tumour was exposed by reflecting the cheek.

Its outer wall was removed with bone forceps, and a cavity, about an inch and a quarter in diameter, and filled with a pale pinkish fleshy growth, was exposed. The growth was scooped out and was not adherent to the walls, which were smooth and glistening.

The cavity was plugged.

The subsequent progress of the case was perfectly satisfactory.

Mr. Mudge, of Hayle, Cornwall, the patient's medical attendant, has kindly informed me that

when last heard of, more than two years after the operation, the man was in excellent health, and there was no return of the tumour.

The minute structure of this tumour presents many interesting features. It was composed almost entirely of small compressed or angular epithelium, in parts (as shown in the accompanying rough sketch) arranged in anastomosing bands which often enclosed rounded spaces, or terminated in club-shaped ends (Fig. VII). Occupying these spaces, and sparsely distributed through the tumour, was a gelatinous fibrillar connective tissue containing branched and round cells. In one section I observed two parallel bands of very large elongated columnar epithelial cells, separated by a narrow band of epithelium having the peculiar characters of those forming the central layer of the enamel organ. (See Fig. VIII*a*.) Scattered at wide intervals were bands of curiously modified connective tissue composed of long clearly defined slender fibrils, and lines formed by the juxtaposition end to end of excessively slender and much elongated fusiform cells. The fibrils and lines were parallel and separated by a clear ground substance. (See Fig. VIII*b*.) There was no evidence of cyst formation in the tumour, and but slight traces of degeneration of epithelium. The relation of the various tissues composing the tumour to those of a rudimentary tooth will be, I think, at once apparent

from the accompanying sketches. Both the form and mode of growth of the epithelium is suggestive of the enamel rudiment, and the solitary band of columnar cells with the intervening network can scarcely be regarded otherwise than as an evolution of the epithelium in the direction of enamel formation. The gelatinous connective tissue likewise suggests the structure of the tooth papilla; while the bands of fibrils may be considered as a modification of connective tissue in the direction of the formation of dentine. If I am correct in believing this to be the case, the specimen throws some light on the mode of formation of dentine, for the fibrils are clearly the product of elongated fusiform connective tissue cells, the intervening substance being of the nature of an intercellular matrix. This seems the more probable as it places the formation of dentine on the same lines as that of bone; the bone lacunæ, and the dentine fibrils are both modifications of connective-tissue cells, and the ground substance is produced by calcification or dentification of the intercellular substance.

CASE 2.—I may here briefly allude to a tumour which was not encapsuled, but which had, in its minute structure, some relation to the preceding. It consisted of round and spindle-shaped nuclei in a homogeneous or finely fibrillar connective tissue, and scattered throughout it were irregular masses and rods of round epithelium, surrounded

by a layer of elongated or columnar cells. The specimen was a very large tumour, involving two-thirds of the lower jaw, and was originally described as a sarcoma. (See Royal College of Surgeons' Museum, No. 2234). It was removed by Mr. Heath from a man aged thirty-two, who eleven years before presenting himself for treatment, noticed a small hard swelling just below the right canine tooth. It remained stationary for five or six years, and began to grow rapidly after a violent blow on the face.*

CASE 3.—This specimen† was sent to me by Mr. T. Smith, by whom it was removed. It comprises a portion of the right side of a lower jaw in the region of the bicuspid teeth. The walls of the bone are expanded by a firm rounded tumour an inch in diameter, having a smooth homogeneous section. The smooth internal surface of the tumour was separated from its bony capsule except above, where it was firmly attached to an irregular mass of osseous substance, which projected downwards from the alveolar surface of the cyst. On closer examination, a small rounded nodule of enamel was found attached to this mass, and may be considered the aborted crown of the rudimentary papilla from which the tumour sprang. The substance of the tumour protruded through an open

* This case is noticed in my lecture above referred to. See Fig. III.

† Preserved in Royal College of Surgeons' Museum, No. 2233.

alveolus, from which the first bicuspid had been removed, after becoming loose. A tooth, probably the displaced first molar, was divided in the operation near the lower part of the cyst, in the wall of which it was firmly embedded. The canine, although inclined backwards, occupied its normal position.

Microscopically the tumour was composed of round and elongated cells, in places undergoing development into fibrous tissue. A layer of elongated cells, resembling odontoblasts, and arranged vertically along the surface, were observed at one part of the tumour.

Mr. Smith has kindly informed me that the patient was a boy, aged fifteen. The tumour was enlarging, but there was no pain; and no history of injury was obtainable.

Perfectly encysted fibro-sarcomatous and fibrous tumours of the jaws do not appear to be of common occurrence. There are two or three typical specimens in the Museum of the Royal College of Surgeons. Mr. Heath* has only observed one case, and he doubts the existence of the embryoplastic odontomata as a distinct class. Broca met with an example in which partial dentification had taken place on the surface of an encysted fibroma. Duplay† has recorded a case in a girl aged twelve,

* *Op. cit.*, p. 267.

† *Op. cit.*, p. 313.

in many respects closely resembling Case 3. A tumour of two years' duration, and occupying the situation of the right incisor and canine teeth of the upper jaw, was destroyed with the thermo-cautery. It returned in three months, and on removal was found to be an encapsuled fibroma of the size of a hazel nut. The incompletely formed crown of a tooth, presenting a resemblance to the canine, projected from the lower and outer surface of the tumour opposite its point of attachment to the cyst wall.

It remains now to consider the relation which the tumours in the cases related bear to other tumours of the jaws and to each other. The relations of the first case of epithelial tumour to the multilocular cystic epithelial tumours of the jaws is undoubtedly close, but there are some important structural and clinical differences. The solid tumour was distinctly circumscribed and non-infiltrating; its structure was more complex, apparently including all the tissues of a rudimentary tooth, and showing no trace of cyst-formation. Clinically there was the significant fact that it ceased to enlarge after the age of twenty-one, a peculiarity shared by other highly organised tumours, as exostoses and digital enchondromata.

Case 3 and Duplay's case present but slight clinical differences from a very large class of more or less completely encapsuled sarcomata of the

jaw in which the distinguished feature of these tumours, namely, the presence of definite dental structures, is wanting; and in Case 2, which differs structurally from many odonto-plastic odontomes merely in the circumstance that its tissues are uncalcified, we have all the characters of an infiltrating malignant tumour.

It is therefore evident that the tumours of which these four cases are examples shade off almost imperceptibly into the general class of tumours affecting the jaws on the one hand, and on the other their affinities with the highly organised odontomata are so close that it seems convenient to retain for them the names of embryo- and odonto-plastic odontomata. In Case 1 no teeth were absent; in Case 3 no sufficiently definite information existed, but it was stated that no teeth were missing; and in Duplay's case the fact that the tumour occupied a position anterior to the molar region renders it probable that the pointed form of a supernumerary cusp was mistaken for that of the canine, therefore the evidence that these tumours originated from the rudiments of milk or permanent teeth is wanting, or inconclusive. And it may be questioned if in some other recorded cases of odonto-plastic odontoma the evidence that the tumour proceeded from the rudiment of some one tooth has not often been forced. Further, it appears unnecessary to

assume that these tumours necessarily spring from the rudiments of aborted teeth, for, as in the case of the multilocular cystic tumours, they may owe their peculiarities in structure to the tooth-forming properties of the tissue in which they arise, whether they spring from the remains of latent embryonic tissue and rudiments of suppressed teeth, or from mature tissues stimulated to active growth by chronic hyperæmia the result of irritation or injury.

If I may be permitted to offer an opinion on a subject with which you must necessarily be better acquainted than myself, I would suggest that from the point of view of general pathology, Broca's class of odontomata do not form a homogeneous group, but includes types of all pathological formations, from the most rudimentary sarcoma up to the true hypertrophy; that the odontomes coronaire and radicaire may rather be regarded as forms of hypertrophy, or giant growths of teeth, than as true tumours, to which class, as a modified variety, the odontomes embryo-plastique and odonto-plastique undoubtedly belong.

In conclusion, I beg to thank you for your kind attention to these, I fear, dry details, and shall consider any trouble I may have taken in the preparation of this paper amply repaid if further facts can be gathered in regard to these peculiar tumours, which lie on the border-land

between tumours generally and odontomata, and, especially, facts relative to their connection or otherwise with the rudiments of normal teeth.

DESCRIPTION OF PLATES.

Fig. I.—Colloid degeneration with the formation of “signet-ring” cells, from the central cells of the columns and alveoli composing the multilocular cystic epithelial tumours. Magnified 650 diameters.

Figs. II, III, IV.—Process of vacuolation and degeneration of epithelium in the above-mentioned tumours, giving rise to a pseudo-reticular tissue. Magnified 650 diameters.

Fig. V.—A portion of the middle layer of a normal enamel-organ showing signet-ring cells and vacuolation of the protoplasm. Magnified 650 diameters.

Fig. VI.—Semi-diagrammatic sketch of a vertical section of the gum at the margin of an open alveolus overlying a multilocular cystic tumour,—to show an ingrowth from the epithelium by which the tumour apparently originated.

a. Margin of alveolus.

b. Ingrowths of epithelium.

c. Alveoli containing pseudo-reticulum and lined with epithelium. Low power.

Fig. VII.—Sketch of solid encysted epithelial tumour (Case I), showing anastomosing bands of epithelium enclosing rounded spaces, with loose fibrillar connective tissue in the interstices of the epithelial structures. Drawn with Obj:D. Zeiss.

Fig. VIII.—From the same specimen as Fig. VII.

a. Portion of parallel rows of enamel-like cells.

b. Dentine-like matrix showing delicate fibrils formed by the apposition end to end, and the elongation of spindle-cells. Magnified 650 diameters.

